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Published in:
Child Care Health and Development

One month after diagnosis: quality of life, coping and previous functioning in siblings of children with cancer[1]

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Accepted for publication 16 April 2004

Abstract

Background The aim of the present study is to describe the quality of life (QoL) of siblings of children with cancer and to predict it according to their health before the diagnosis of cancer in the ill child and their ways of coping with the illness.

Methods Participants were 83 siblings from 56 families – 46 girls and 37 boys, aged 7–18. The assessment took place 1 month to 8 weeks after the diagnosis of cancer in the ill child. The siblings' QoL was assessed with the TNO-AZL children's quality of life questionnaire (TACQOL). Coping strategies were assessed with the Cognitive Coping Strategies Scale for siblings (CCSS-s). Physical problems and eating and sleeping problems that existed before the ill child was diagnosed were determined in a structured interview with the parents.

Results A substantial number of siblings reported impaired cognitive and emotional QoL compared to the reference group. School-aged siblings (7–11 years) reported more trouble with motor functioning than peers. The coping strategy 'predictive control' (maintaining positive expectations regarding the illness) positively predicted siblings' QoL. The presence of health problems before diagnosis was negatively associated with siblings' QoL. Older siblings reported more negative emotions, while girls reported lower social QoL and reliance on 'interpretative control' (trying to understand the illness) was associated with fewer positive emotions.

Conclusions During the first 2 months after the diagnosis of cancer in a brother or sister, siblings have relatively lower QoL than peers. Health problems that existed before diagnosis may be a predictor of later adjustment problems. Positive expectations about the course of the illness appear to protect siblings from distress. Information about the illness is a delicate issue that requires parental guidance.

Keywords
paediatric cancer, siblings, quality of life, coping, psychosocial functioning

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Introduction

When a child is diagnosed with cancer, the whole family must face an extraordinarily stressful phase. The medical treatment that the ill child must undergo causes the family life to change drastically. The diagnosis of a life-threatening disease, the unpredictable course of the illness, and feelings of

[1] The present study results have been published in Dutch, in the *Tijdschrift Kindergeneeskunde* (Dutch Journal of Pediatrics), 71 (3), 2003, 123–133.

uncertainty and helplessness that can result from the inability to exert control over the illness, can cause intense emotional reactions in family members, including the siblings of the ill child. How siblings of children with cancer adjust to the illness of their brother or sister has been studied more and more systematically during the last 20 years (Houtzager *et al*. 1999).

In several studies the results indicated that siblings of children with cancer are at risk for emotional, internalizing problems and/or behavioural, externalizing problems (Spinetta 1981; Schuler *et al*. 1985; Fife *et al*. 1987; Walker 1988; Bendor 1990; Carpenter & Sahler 1991; Cohen *et al*. 1994; Barbarin 1995; Heffernan & Zanelli 1997; Packman *et al*. 1997). Besides, there are indications that siblings of children with cancer have lower social competence compared to peers (Evans *et al*. 1992; Cohen *et al*. 1994). Given the enormous changes in daily routines and in family life that siblings of a child with cancer must undergo, siblings may be affected on other domains than merely the behavioural and emotional ones that have been investigated. Siblings' overall quality of life (QoL) may very likely be affected as well. In paediatric research, much attention is paid to the effect of treatment on the child's QoL in order to be able to evaluate medical treatment, develop new protocols, and provide focused psychosocial care where needed. However, the effect of treatment extends beyond the ill child and, as demonstrated above, can affect siblings' well-being. It is therefore all the more remarkable that the effect of paediatric cancer on siblings' QoL has not been investigated yet. Questionnaires that measure QoL may be more sensitive to sibling adjustment problems, because they are not focused on determining the presence of psychopathology, but on the evaluation of a child's quality of overall well-being. The aim of the present study is therefore to assess to what extent siblings' QoL is affected by the illness of their brother or sister. Another goal is to investigate how it can be predicted according to siblings' previous functioning and coping efforts.

Not all siblings are equally affected by the illness. For clinical practice it is essential to understand why some siblings function relatively well, despite the illness, whereas others are vulnerable for adjustment problems from diagnosis onwards. Specific psychosocial problems that are present in the sibling before the onset of the illness may be a sign of vulnerability. Ideally, such problems should be recognized early in the disease process. Kalnins *et al*. (1980) stated that the disease of a family member not only causes new disease-related problems, but that pre-existing problems in the psychological and social areas deteriorate in many cases. This relationship was demonstrated in a quantitative US study (Sahler *et al*. 1994). A substantial number of children (40%) who had emotional or behavioural problems before the diagnosis of cancer in a sibling had serious psychosocial adjustment problems after the diagnosis that would require psychosocial care. On the other hand, only 2% of the children without any emotional or behavioural problems before the diagnosis were found to have serious psychological problems after the diagnosis. Problematic social functioning within the family system may be predictive of later adjustment problems as well. Fife *et al*. (1987) found an association between the quality of the relationship between parents and children before the diagnosis on the one hand, and the occurrence of psychosocial problems after diagnosis on the other hand. These studies demonstrate that siblings with psychosocial problems before the diagnosis may be at risk for adjustment problems as the illness progresses. Although such problems are difficult to recognize, their determination is essential for the development of early interventions and prevention of adjustment problems in siblings.

The next step is to enhance the competences and resources of these vulnerable siblings. But what resources need to be the focus of interventions? The literature on psychosocial adjustment of siblings to paediatric cancer still provides inconsistent information on resources that may protect children from psychosocial adjustment problems (Chesler *et al*. 1992; Barbarin 1995; Sloper & While 1996). Knowledge on effective coping strategies that help siblings adjust positively to the illness may provide indications on how to prevent or treat psychosocial problems in siblings at risk. However, given the fact that coping still has received too little systematic attention in paediatric research in gen-

eral (De Ridder & Schreurs 2001), knowledge on how siblings of a paediatric cancer patient cope with the illness is even scarcer. Of 35 studies reviewed, only two had used standardized questionnaires to investigate how siblings of paediatric cancer patients cope with the illness (Houtzager *et al.* 1999). In order to be investigated properly, the concept of coping requires further definition. The most broadly cited model of stress and coping applied in most research is that of Lazarus and Folkman (1984). According to Lazarus and Folkman (1984), coping refers to all individual actions that are focused on handling stressful situations. The basic assumptions to the stress and coping model of Lazarus and Folkman are, that coping is a process, that there is no universally 'good' or 'bad' coping but that it depends on numerous individual and situational aspects, and that outcome is adaptation instead of psychopathology. Furthermore, coping resources should be evaluated separately from adjustment outcome in order to truly measure their adaptiveness (Lazarus & Folkman 1984; Lazarus 1993). Coping is usually categorized into problem-focused and emotion-focused strategies. On the one hand, an individual can undertake action to solve the problem that causes the stressful situation (problem-focused coping). On the other hand one can try to reduce unpleasant feelings that were induced by the situation, by cognitive or emotion-focused actions (emotion-focused coping). Both ways of coping can occur separately or simultaneously in the same situation. Kliewer and Sandler (1992) argue that the nature of the stressful situation determines which way(s) of coping are effective in dealing with that situation. It has been demonstrated in several studies that children's coping strategies should match the situation in which they find themselves, in order to be effective (Boekaerts & Roder 1999). In situations that are uncontrollable, strategies that are directed at redefining the situation, in order to be less threatening (secondary coping strategies), may be more effective in reducing of feelings of distress (Rothbaum *et al.* 1982). As childhood cancer is mostly beyond the control of the cancer patient, and in this case, family members, problem-focused coping strategies may occur least often (Lazarus & Folkman 1984). Both the ill child and his or her parents and siblings therefore may be most likely to rely on ways of coping that are focused on resisting the threat and reducing, as much as possible, the unpleasant feelings that may come up. Cognitive or emotion-focused coping can therefore be assumed to occur most often in siblings of paediatric cancer patients. The two-process model of perceived control of Rothbaum *et al.* (1982), in which such emotion-focused or 'secondary' coping strategies were defined, has showed to be a useful approach in understanding the adjustment of children with cancer and their parents because it takes into account the uncontrollability of the situation (Grootenhuis *et al.* 1996; Grootenhuis & Last 2001). By trusting the expertise of the medical staff or by maintaining an optimistic view on the disease process, by trusting in higher powers or by efforts directed at understanding the disease, for instance, family members can reduce their negative feelings. These are examples of secondary control strategies; ways of thinking about the disease by which negative and threatening emotions can be dealt with. Grootenhuis and Last (2001) showed that the emotional well-being is influenced by the extent to which children with cancer rely on cognitive coping strategies. Paediatric cancer patients who managed to maintain a positive and optimistic attitude towards their illness were reported to experience lower levels of fear and fewer depressive symptoms. One of the research questions addressed here is to what extent these cognitive coping strategies are applicable to the psychosocial adjustment of the patient's sibling(s). Four secondary control strategies that are based on Rothbaum's model, further referred to as 'cognitive control strategies', will be investigated in the current study: predictive, vicarious, interpretative and illusory control. *Predictive control* refers to optimism, or efforts aimed at maintaining a positive outlook on the illness. This cognitive coping strategy may enable siblings to increase the predictability of the situation, thus enhancing feelings of control. *Vicarious control* refers to the perception that others, such as the medical staff, can exert control. Putting trust in the medical treatment and capabilities of the doctor may relieve feelings of uncontrollability. *Interpretative control* is represented by efforts to

understand the illness and derive meaning from it. Enhanced understanding of the illness may reduce feelings of uncertainty and increase its predictability. *Illusory control* refers to wishful thinking or hoping for a miracle. Belief in a greater power or the possibility of a miracle may reduce feelings of helplessness and may provide hope.

The purpose of the present study is threefold. Firstly, this study was aimed at the assessment of siblings of paediatric cancer patients' QoL. For this purpose, the sibling's QoL shortly after the diagnosis of paediatric cancer in a brother or sister will be compared with available reference groups of children the same age. A second purpose of this study is to determine how vulnerable children can be recognized at an early stage. Therefore, the siblings' previous health will be assessed and related to their post-diagnosis QoL. Previous health will be conceptualized as the presence of common health problems, such as eating, sleeping and physical problems that are relatively easy to observe and to determine retrospectively. Thirdly, the reliance on the cognitive coping strategies of predictive, vicarious, interpretative and illusory control are assessed in association with siblings' QoL. The results of the current study may reveal to what extent QoL questionnaires are applicable for determining the impact of paediatric cancer on siblings. Furthermore, information on the role of health problems before the diagnosis and subsequent reliance on cognitive coping strategies can enable early recognition of adjustment problems and can guide focused interventions.

Methods

Procedure

From December 1998 to January 2001, parents and siblings of children treated in the paediatric oncology ward in the Emma Children's Hospital of the Academic Medical Centre in Amsterdam were asked to participate in the present study. From July 2000 onwards, families of children treated in the paediatric oncology ward of the University Hospital in Groningen were recruited as well. Inclusion criteria were that: the ill child was diagnosed with a malignant tumour no more than 4 weeks ago; the sibling(s) was (were) aged 7–18 years old; and parent and sibling had sufficient command of the Dutch language. To account for over-representation of large families, no more than two siblings from each family were included in the study.

Participants

Of 71 eligible families, 56 (78.9%) agreed to participate (Table 1). The study group thus consisted of 83 siblings, 46 girls and 37 boys aged 7–18, with a mean age of 11 (SD = 3). No significant differences were found between the study group and the non-response group regarding age, gender and diagnosis of the ill child, and age and gender of the siblings. After informed consent was obtained, an appointment was made to visit the family. Questionnaires were sent in advance, with explicit instructions to complete them independently and individually. Two psychologists visited the family. They collected the questionnaires and conducted interviews with the parents. If necessary, younger siblings were assisted with the completion of questionnaires. The assistance of the psychologist was restricted to reading out loud questions and explaining of the meaning of difficult words. The assistance took place in a separate room, in the absence of the sibling's parents.

Table 1. Characteristics of participating siblings and families

	n	%
Families included	56	78.9
Siblings	83	
Gender		
Male	37	44.6
Female	46	55.4
Age (year)		
7–12	59	71.1
13–18	24	28.9
Ill child	56	
Diagnosis		
Leukaemia	13	23.2
Lymphoma	13	23.2
Solid tumour	25	44.6
Brain tumour	5	8.9
Parents	102	
Fathers	49	48.0
Mothers	53	52.0

Measurements

Health-related QoL was assessed with the TNO-AZL children's quality of life questionnaire (TACQOL) (Vogels *et al*. 2000) for children aged 8–15. Health-related QoL is defined as health status (HS) in combination with a health-related (HR) component, which represents the emotional evaluation of one's HS. The TACQOL consists of seven scales, with eight items each: body, motor, autonomous, cognitive functioning, social functioning, positive emotions and negative emotions. Except for positive and negative emotions, each item is stated as a functional problem. The child has to indicate whether a particular problem is present, and how he or she feels about the problem, the HR component. Items are scored as follows: not present (0), if present, the child feels: good (1), not so good (2), quite bad (3) and bad (4) about this particular problem. These items have a maximum score of 32, with higher scores representing better health-related QoL. The occurrence of positive and negative emotions, such as happiness, cheerfulness, sadness, or anger, are scored on a three-point scale: often (0), sometimes (1) and never (2), resulting in a maximum score of 16, with higher scores representing a more favourable emotional QoL. Healthy reference groups of children aged 8–16 are available for the TACQOL (Vogels *et al*. 2000).

The siblings' reliance on cognitive coping strategies was assessed with the Cognitive Coping Strategies Scale for siblings (CCSS-s). The CCSS is an illness-specific self-report questionnaire that was developed for use with children that grow up with a chronic or life-threatening disease. The CCSS assesses to what extent children try to maintain a sense of cognitive control, by relying on cognitive coping strategies (Grootenhuis *et al*. 1996; Grootenhuis & Last 2001). Four cognitive coping strategies are assessed: predictive control, vicarious control, interpretative control and illusory control. These four strategies represent the reliance on different ways of cognitive coping. The questionnaire for children with a chronic or life-threatening illness (Grootenhuis *et al*. 1996; Grootenhuis & Last 2001) was used in order to develop the sibling version of this questionnaire. The statements of the patient questionnaire were converted to suit the situation of siblings. For example, the statement 'I worry about my disease' was converted into the statement: 'I worry about my brother or sister's disease'. The sibling version of the CCSS was first tested in a pilot group of siblings. Then, scales were constructed with use of factor analysis. The final version of the sibling questionnaire consists of 20 statements, on which siblings can indicate on a four-point Likert scale to what extent they agree (totally agree, agree, disagree and totally disagree). Four items were omitted after factor analyses, resulting in the subscales: predictive control (four items), vicarious control (six items), interpretative control (four items), and illusory control (two items). Internal consistency (Cronbach's alpha) of these scales is 0.81, 0.70, 0.71 and 0.30, respectively. Given its low reliability, the siblings' reliance on illusory control does not seem to be relevant for siblings. The illusory control scale will therefore be excluded from further analyses.

Sibling health before onset of the illness in their brother or sister was assessed retrospectively in a structured interview with the parents. To enhance the reliability of this retrospective measurement, parents were asked to recall relatively observable problems in daily functioning. Problems were assessed in three domains: physical problems (headaches, stomachaches, nausea, or other unspecified physical complaints), problems sleeping (problems falling asleep, remaining asleep, nightmares, enuresis or other unspecified sleeping problems), and eating problems (over- or under-eating, being fuzzy about food, or other unspecified eating problems). The research psychologist asked the parent to report whether any of these problems had occurred during the 2 months preceding the diagnosis. For each specific complaint, the frequency of occurrence was assessed from (0) never, (1) monthly, (2) weekly, to (3) daily. Scores of physical complaints ranged from 0 to 6 (Mean = 0.82; SD = 1.42); sleeping problems ranged from 0 to 6 as well (Mean = 0.83; SD = 1.61) and eating problems ranged from 0 to 3 (Mean = 0.51; SD = 1.06). Previous health was converted into a total score of previous health that ranged from 0 (no complaints, $n = 39$) to 11 (Mean = 2.2; SD = 2.9) and was used in the present analyses.

Statistics

Before differences between siblings and the reference group could be assessed, we needed to investigate whether QoL scores of the siblings that were not represented in the reference group (siblings aged 7 and 17–18) differed from those of the other siblings. Scores of siblings aged 7 were compared to those of siblings aged 8–11, and scores of 17–18 years older siblings were compared to those aged 12–16. No differences were found with the other age group for siblings aged 7 ($n = 5$). Adolescent siblings aged 17–18, however, reported lower QoL on physical functioning ($n = 5$; Mean = 16.6), compared to siblings aged 12–16 ($n = 29$; Mean = 23.3) ($t = 2.92$; $P = 0.006$). On the other subscales of QoL, no differences were found between those two age groups. Considering their low QoL scores on physical functioning, siblings older than 16 were excluded from the analyses for this subscale.

Mean differences between siblings and the reference group on subscales of QoL were assessed, using Student's *t*-tests. Analyses were performed for younger siblings (7–11 years old) and adolescent siblings (12–18 years old) separately. Mean scores on various domains give an indication of how siblings of children with cancer are doing. The question of prevalence of serious adjustment problems that may require further psychosocial care remains unanswered when the focus is on mean scores. Besides, differences in the distribution of scores between the study group and the reference group are problematic, when mean scores are compared. Ceiling effects and skewed, non-normal distributions are a recognized problem in assessment of QoL (Landgraf *et al.* 1998; Rose *et al.* 1999). One solution to this problem is to investigate how large the risk group with seriously affected QoL is. For this purpose, a borderline was determined at the 20th percentile in the healthy reference group with children of the same age and gender. Scores below this borderline thus correspond with the lowest, most 'affected' QoL scores in the normal population, represented by the reference group. If the incidence of impaired QoL in siblings is comparable to the normal population, 20% of the study group will have scores in this impaired QoL range. Differences in the distribution of 'affected' and 'non-affected' children between our study group and the reference group were assessed using chi-square tests. Because of low variance on the autonomy scale, results will not be presented for this domain.

Subsequently, the association of QoL with coping and previous functioning was assessed. Repeated simultaneous regression analyses were conducted for all domains of QoL with sibling age and gender, their reliance on cognitive coping strategies, and sibling previous functioning. A forward stepwise procedure was conducted. Predictors were selected if their contribution to the variance in QoL scores was significant, with a *P*-value of less than 0.05. Selection continued until variables that had a significant contribution to the model remained.

Results

Complaints before diagnosis

In 52% of the siblings in the study group parents reported health complaints with regard to sleeping, eating, and physical functioning in the 2 months preceding the diagnosis of cancer in the ill child. About one-third were reported to have physical complaints like headaches, stomachaches, sickness or other symptoms. Just under a third of the siblings were reported to suffer from sleeping difficulties and one-fifth had eating problems (Table 2).

Quality of life in comparison with the reference group

Siblings of 7–11 years old reported a lower QoL than the reference group in the following domains: motor skills, cognitive skills, positive and negative

Table 2. Prevalence of health complaints in siblings before the diagnosis

	Total	Physical complaints	Sleeping problems	Eating problems
Children with one or more health complaints				
%	52.4	35.4	26.8	20.7
n	43	29	22	17
Number of reported complaints				
Mean	2.16	0.82	0.83	0.51
(SD)	(2.92)	(1.42)	(1.61)	(1.06)

Table 3. Self-reported quality of life (means) in siblings compared to reference data

				Reference group		
TACQOL† domains	***n***	**Mean**	**SD**	***n***	**Mean**	**SD**
Siblings aged 7–11 years						
Physical functioning	44	24.3	5.9	896	25.3	5.0
Motor functioning	45	29.0*	3.8	895	30.0	3.1
Autonomy	45	30.8	2.6	896	31.3	1.8
Cognitive functioning	45	27.2*	4.5	894	28.6	3.9
Social functioning	43	29.0	3.8	892	29.8	2.6
Positive emotions	44	11.7***	3.1	884	13.6	2.5
Negative emotions	44	10.6**	2.7	883	11.7	2.7
Siblings aged 12–18 years						
Physical functioning	29	23.4	4.8	824	24.8	5.0
Motor functioning	35	29.4	3.3	826	30.1	3.1
Autonomy	35	31.5	1.7	825	31.6	1.3
Cognitive functioning	35	26.2*	4.9	826	27.9	4.0
Social functioning	35	28.3	4.7	823	29.4	3.1
Positive emotions	35	10.4***	3.3	819	13.1	2.8
Negative emotions	35	9.7***	3.2	819	11.8	2.5

*$P < 0.05$, **$P < 0.01$, ***$P < 0.001$.

†TACQOL, TNO-AZL children's quality of life questionnaire. Higher scores represent a more favourable quality of life, indicating that fewer problems are reported.

emotions (Table 3). Lower QoL on motor skills refers to the negative evaluation of limitations in walking, running, or energy. Lower cognitive QoL refers to the negative evaluation of problems concentrating and difficulties with memory and learning. A lower QoL in the negative emotional domain refers to experienced feelings of jealousy, anger, aggression, sadness, worry, gloominess, fear or depression, whereas a lower QoL on positive emotions refers to the relative absence of feelings like happiness, joy, satisfaction, relaxation, enthusiasm and cheerfulness. Like the younger siblings, adolescent siblings aged 12–18 experienced a lower QoL than peers regarding cognitive skills, positive and negative emotions. Their evaluation of motor skills was comparable to the norm (Table 3).

Sibling quality of life

In the present study, impaired QoL was assumed when scores were below the 20th percentile in the reference group. The risk area is determined on the basis of the sibling's gender and age (7–11 and 12–18 years old). The results are displayed in Fig. 1 for the following areas: physical functioning, motor skills, cognitive functioning, social functioning, positive emotions, and negative emotions.

Impaired QoL was reported by 26–56% of the siblings. The prevalence of impaired QoL is significantly higher than in the reference group, in the whole group and in both age groups separately (Fig. 1). Physical complaints (31%), problems with motor skills (32%), problems with cognitive abilities (37%), social problems (33%) and negative emotions (33%), occurred relatively more often than in the reference group (less than 20%). More than half of the younger siblings (56.6%) and adolescent siblings (57%) indicated that they experienced few positive emotions. Besides, a large subgroup of adolescent siblings (40%) reported having elevated negative emotions.

Predictors of quality of life

In Table 4 the association of age and gender, reliance on cognitive coping strategies, and health problems before the diagnosis with siblings' QoL is displayed. Firstly, sibling age was positively related to negative emotions, and sibling gender was related to social QoL. With increasing age, siblings reported significantly more negative emotions. As to gender, female siblings reported more problems on the social domain of QoL than male siblings, indicating that they are less satisfied with relationships with family members, peers and others.

Two of the investigated cognitive control strategies were associated with QoL. Predictive control was positively associated with cognitive and emo-

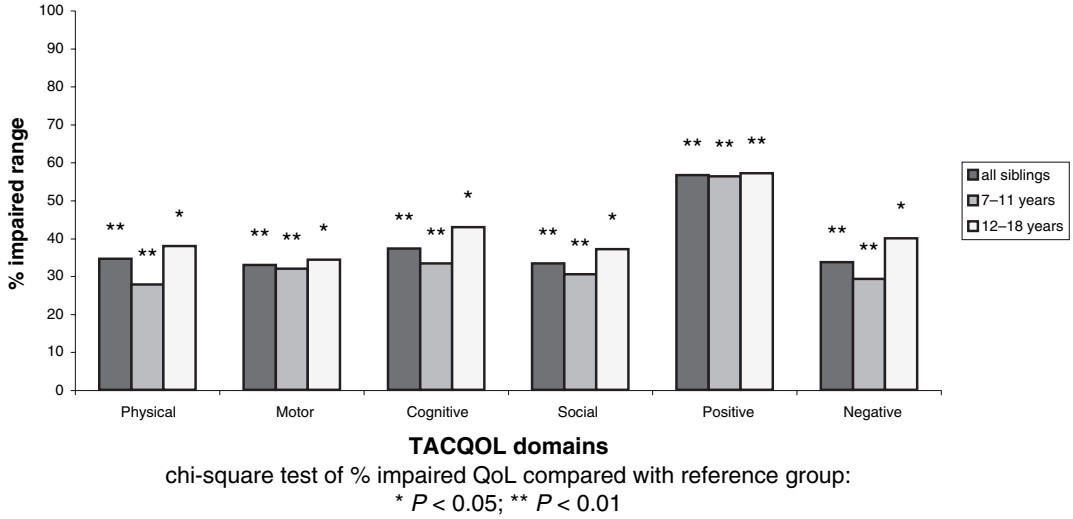

Figure 1. Percentage of siblings with scores in the 20th percentile of impaired quality of life according to reference data. TACQOL, TNO-AZL children's quality of life questionnaire.

Table 4 Predictors of sibling quality of life: age, gender, cognitive coping strategies and health problems before diagnosis (stepwise regression analysis)†

	TACQOL‡ domains						
Model	**Physical functioning**	**Motor functioning**	**Autonomy**	**Cognitive functioning**	**Social functioning**	**Positive emotions**	**Negative emotions**
R^2	0.04	0.08	0.13	0.16	0.08	0.14	0.18
F	3.11	6.78*	1.75	6.87**	6.69*	6.00**	5.42**
Significance of *F* (*P*)	0.08	0.011	0.123	0.002	0.012	0.004	0.002
Demographics							
Age							–0.24*
Gender					–0.28*		
Control strategies							
Predictive control				0.25*		0.33**	0.23*
Vicarious control							
Interpretative control						–0.29*	
Previous functioning							
Health complaints before diagnosis		–0.28*		–0.33**			–0.26*

Significance of *F* and Beta: *$P < 0.05$, **$P < 0.01$.

†Regression coefficients (betas) are displayed. Regression coefficients were included only when a significant contribution to the regression model was obtained.

‡TACQOL, TNO-AZL children's quality of life questionnaire. Higher scores represent a more favourable quality of life, indicating that fewer problems are reported.

tional QoL, and interpretative control was negatively related to positive emotional QoL. This means that, the more siblings are able to rely on positive expectations regarding their sibling's illness (more predictive control), the better their cognitive functioning and the more positive and the fewer negative emotions they experience. Besides, siblings reported fewer positive emotions when they were more inclined to try to find explanations for the illness or to try to understand the illness (more interpretative cognitive control). The occurrence of health problems previously to the diagnosis in the ill child was related to siblings' QoL after the diagnosis. Siblings who were reported to have

had many physical, sleeping and eating problems before the diagnosis, reported more problematic motor and cognitive functioning, and reported more negative emotions.

Discussion

The purpose of the current study was to assess in which areas and to what extent QoL of siblings of children with cancer is affected shortly after the diagnosis in the ill brother or sister. In addition, the present study aimed to investigate whether children with previously existing functional complaints were more vulnerable after diagnosis and whether the reliance on cognitive coping strategies enabled siblings to adjust more favourably to the illness shortly after the diagnosis.

The results revealed that there is a considerable subgroup of siblings who report difficulties in a number of areas. Siblings aged 7–11 and 12–18 years experience more cognitive and especially more emotional problems than their peers. Siblings aged 7–11 years reported more motor difficulties than peers. TACQOL motor skills difficulties refer to problems in the areas of walking, standing, running, walking stairs, balance, playing, speed and stamina. Parallel to these findings, a higher incidence of somatic problems on the Child Behaviour Checklist (CBCL) was found in siblings of children with cancer in an US study (Zeltzer *et al*. 1996). It is understandable that emotionally affected siblings feel apathetic and therefore may have relatively less energy and greater difficulties putting in physical effort. Low QoL scores on motor skills therefore may reflect low energy levels resulting from the distress siblings experience shortly after diagnosis, or otherwise these scores may be an expression of worries about their own health.

The reported differences between siblings of children with cancer and their peers demand reference to their clinical relevance. QoL scores were therefore divided into a 'risk' and a 'healthy' group. Siblings in the risk group had scores below the 20th percentile norm. In all areas of QoL, a significant number of siblings reported scores within the risk group. Approximately 50% of the children experience significant emotional difficulties, an incidence that was significantly higher, and more than twice as high compared to the reference group. Nearly 50% of the siblings reported excessive negative feelings such as anger, sadness, worry, gloom, jealousy and fear. A majority of the siblings reported that positive feelings such as feeling cheerful, lighthearted, content, enthusiastic, relaxed, happy, self-confident, or glad, were relatively absent in comparison with peers.

The investigated characteristics and resources of the siblings were associated with different aspects of their QoL. Firstly, there was an association with age and gender. Adolescent siblings appeared to be more vulnerable for a negative mood. The results also revealed that girls were particularly at risk for difficulties in their social relationships. In two other studies, sisters have been reported to be particularly at risk for emotional and behavioural problems (Sahler *et al*. 1994; Packman *et al*. 1997). The authors suggested that parents might expect teenaged sisters to take on relatively more responsibilities in the care for the ill child. As a result, girls may be more restricted in social activities outside the home and, consequently, may become isolated from their friends than male siblings. Secondly, the reliance on cognitive coping resources was associated with siblings' QoL. Siblings' reliance on predictive control was associated with higher cognitive and emotional QoL. This means that siblings who were able to maintain positive expectations regarding the illness and who remained optimistic (predictive cognitive coping), adjusted more favourably to the illness. In a previous study on the importance of cognitive control strategies it became evident that predictive control was an indicator for lower levels of anxiety and fewer depressive symptoms in children who were treated for cancer (Grootenhuis & Last 2001). An optimistic attitude may give children a sense of control and protect them from the negative emotions that are automatically induced by the uncontrollable consequences of the illness of their brother or sister. Besides the effect of predictive control, interpretative control was related to siblings' QoL: Siblings who desire to understand the meaning of the illness and ask questions about their brother or sister's illness (interpretative cognitive control) experience fewer positive emotions. This finding may best be under-

stood in the context of children's coping with stress-inducing information. Siblings that rely highly on interpretative control may be more likely to encounter negative information about the illness than siblings who are not inclined to seek information. Different studies demonstrated that children who tend to focus on threatening information generally respond with more fear than children who do not necessarily seek threatening information. For example, Muris *et al*. (2000) showed a direct relationship between such 'approaching' styles of coping in children and the prevalence of symptoms of fear. Miller *et al*. (1995) studied coping strategies in children aged 7–12 who underwent an invasive dental operation. Children who tended to focus on the threatening aspects of the situation (monitoring) were more frightened. The same mechanism may be responsible for the finding presented here. Siblings who indicated that they wanted to know everything about the disease may expose themselves to anxiety-inducing information about the illness of their brother or sister. If parents are not there to discuss the new information about the illness, this can cause unrealistic fears and therefore make children more vulnerable for distress. Finally, the study results indicated that health complaints before the diagnosis of the ill child may be a sign of pre-diagnosis vulnerability. Physical complaints and eating or sleeping problems predicted the occurrence of motor skills difficulties, cognitive problems and negative emotions.

There are several limitations to the present study that place the aforementioned results into perspective. Firstly, the siblings' QoL was only partly explained by the predictors that were investigated here. Other explanatory factors such as family functioning, illness severity, other ways of coping and other factors may be of importance as well. Besides, interaction of coping with other variables such as the diagnosis and prognosis is not unlikely, however, was not addressed here. The positive association of predictive control (having a positive, optimistic outlook) with QoL could be hypothesized to depend on the ill child's prognosis. However, in a previous study, children with cancer who had experienced a relapse, did not to rely on predictive coping strategies any more or less than other patients (Grootenhuis & Last 2001). Whether this is the same for the patient's siblings will have to be investigated in future studies. Another limitation of the present study concerns the questionnaires that were used. The CCSS for siblings was developed specifically for children who have a sibling diagnosed with cancer. This means that it could not be validated in large groups of peers. However, the researchers did test the questionnaire in a pilot study. The CCSS-s was also based on a questionnaire that is used in paediatric populations. Results with the CCSS in different populations seem to be quite consistent and the present results demonstrated that the concept of cognitive coping is meaningful in this population as well. The advantage of a disease-specific instrument is that it is more sensitive to siblings' experiences, and therefore may generate relatively meaningful results, compared to a generic questionnaire. Previous health problems were not investigated with a standardized questionnaire either. Such problems had been reported in half of the siblings (52%). This incidence could, however, not be compared to reference data. Because these were assessed retrospectively in an interview with the parents, the accuracy depends on the parent's memory that may be biased by the distress they experienced at the time of this assessment. To restrict bias as much as possible, relatively observable symptoms were chosen to be assessed. Another problem with this measurement is that confrontation with illness symptoms in the period before the diagnosis may have induced stress symptoms in the siblings before the actual diagnosis. Although the aforementioned problems should be considered in the interpretation of the findings, the diagnostic and predictive relevance of health problems was apparent. Lastly, the TACQOL was used. Reference data of the TACQOL were limited to children aged 7–16. Because siblings aged 17–18 years were not represented in the reference group, reference data were not completely representative for the present study group. The wide age range in the study group might appear problematic as well. Gender and age differences within a study group are a frequently encountered problem in paediatric research. It results in analyses in small subgroups and requires interpretation of developmental differences between subgroups. The wide age range that was

assessed with the TACQOL was also because of the preliminary status of the questionnaire at the start of this investigation. The TACQOL now has a child version and an adolescent version. Nevertheless, the advantage of the use of one single questionnaire is that a larger data set could be used in the regression analyses.

The present results demonstrated that the determination of QoL in the unaffected healthy children in families with seriously ill children is informative. The term QoL appears to be applicable to these children, because they go through radical changes that are not without consequences. For the medical staff it is important to notice both somatic and psychosocial difficulties in siblings at an early stage and, if necessary, refer the family for further support. The medical staff, and in particular the general practitioner, should be alerted when frequent health problems in siblings precede the diagnosis of the patient. These problems can be a sign of vulnerability for later difficulties. Results also stress the need to advise parents in how and when information about the illness and treatment is given to the siblings of the ill child. Providing siblings with potentially threatening information seems to counteract parents' natural inclination to protect their child against negative emotions, adversities and threat. However, if facts about the illness are covered up, children can become convinced that the disease is too bad to talk about. This prevents the parents and the child from sharing worries and experiences and seeking each other's support. On the other hand, if siblings go and seek information about the illness by themselves, they have no one to reflect upon it and therefore may become more insecure instead of more knowledgeable. Giving information openly can give the child the option to find out, with the parents, what the implications are of the factual information on their brother or sister's disease. It is obvious that the information should be adjusted to suit the age and understanding of the child. For the medical specialist, an important but difficult task emerges from the present results. The oncologist needs to acknowledge the facts of the disease on the one hand, while at the same time leave room for reassurance and optimism. The honest discussion of the illness and treatment creates an atmosphere of confidence and trust that is essential for a good relationship between family members. Sometimes, the siblings may want to join meetings with the oncologist. When parents experience difficulties explaining the illness to siblings, the oncologist can be of great help in providing siblings with balanced information. If adjustment problems appear to be more profound, additional support is needed for siblings. In these cases, attending a support group may be helpful. In such groups, children can receive and discuss well-balanced information and learn how to cope with difficult emotions. Besides, they may feel supported by sharing feelings and experiences. It has been demonstrated that such support groups for siblings can be effective (Houtzager *et al.* 2001). In future studies siblings' QoL certainly needs to be investigated further, in order to validate the present findings. Further research is also needed in order to show how the psychosocial problems found here develop over time as the treatment progresses; and to what extent the previously discussed coping styles could be of long-term influence to the sibling's QoL.

Acknowledgements

We would like to acknowledge the financial support by the Dutch Cancer Society (NKB/KWF) and the Foundation for Paediatric Cancer Research (SKK), which enabled us to carry out this study. We would also like to express many thanks to Astrid Ball for her effort to translate the original text, and to Saskia Vlaming, Marieke de Boer and Daisy Theunissen for their efforts in the data collection and data management.

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